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CASE REPORT

Peritoneal Implantation of Squamous Cell Carcinoma Following Rupture of a Dermoid Cyst during Laparoscopic Removal

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Background. **In recent years, there has been a move toward using laparoscopy in the management of presumed benign ovarian masses. This paper discusses the use of laparoscopy for ovarian masses (particularly dermoid cysts). The focus is on the implications for unexpected malignancies.**

Case. **We report here a case involving a dermoid cyst that was removed at laparoscopy and was subsequently found to contain a squamous cell carcinoma. Spillage of the cyst's contents occurred at the time of removal. At staging laparotomy, peritoneal implants of the tumor were found, upgrading the tumor from FIGO stage Ia to IIc.**

Conclusion. **Spillage rates of dermoid cyst content with laparoscopic removal are inevitably higher than with excision at laparotomy. It remains controversial whether upstaging from FIGO stage Ia to IIc affects prognosis; however, in this case it led to aggressive adjuvant therapy which imposed significant additional morbidity, including loss of fertility, on the patient. One should be aware of the possibility of unexpected malignancy when the decision to manage an ovarian mass laparoscopically is made.**

INTRODUCTION

The benefits of laparoscopy over laparotomy in the management of ovarian cysts include decreased postoperative pain, less time spent in the hospital, and a shorter recovery period [1]. Laparoscopic management of dermoid cysts of the ovary has raised concerns about the associated increased risk of spillage of dermoid contents into the peritoneal cavity. Cases of chemical peritonitis with subsequent extensive adhesion formation and granulomatosis have been reported following intraperitoneal rupture of dermoid cysts; however, other studies have reported the absence of complications with spillage when this was followed by generous irrigation of the abdominal cavity [1, 2]. The risk of spillage of malignant cells following rupture of a dermoid cyst is often minimized because malignancies occur only rarely in such cysts.

We present here a case of stage Ia squamous cell cancer arising in a dermoid cyst of the ovary where intraoperative spillage of the cyst's contents resulted in upstaging to stage IIc.

CASE REPORT

A 37-year-old gravida-2 para-1 aborta-1 patient presented to the emergency department with right lower quadrant pain of acute onset. Examination was significant for the presence of a tender pelvic mass. Ultrasonographic examination revealed an inhomogeneous (cystic and solid areas) 5.0-cm lesion of the right ovary containing echogenic material with dirty shadowing. This was felt to be consistent with a diagnosis of mature cystic teratoma (dermoid cyst) of the ovary.

The patient's past obstetrical and gynecological history was significant for one spontaneous vaginal delivery 8 years previously and an elective induced abortion in the remote past. She had noticed irregular menses in the preceding 12 months. Her past medical history was otherwise unremarkable.

An emergency laparoscopic surgery was arranged for a provisional diagnosis of a dermoid cyst with torsion of the right ovary. The findings at laparoscopy were consistent with a dermoid cyst. However, there was no evidence of torsion or rupture seen to account for the discomfort. The wall of the cyst was smooth. There was no evidence of excrescences or peritoneal implants. Electrocautery with a cutting current was used to incise the dermoid cyst. Contents of the dermoid cyst (hair and sebaceous material) were removed by suction. No attempt was made to isolate the cyst in a bag. There was spillage of the contents of the cyst and irrigation was used to remove spilled contents. The collapsed cystic mass was then removed leaving the right ovary *in situ.* A small left ovarian cyst was also removed during the procedure. The patient recovered from the

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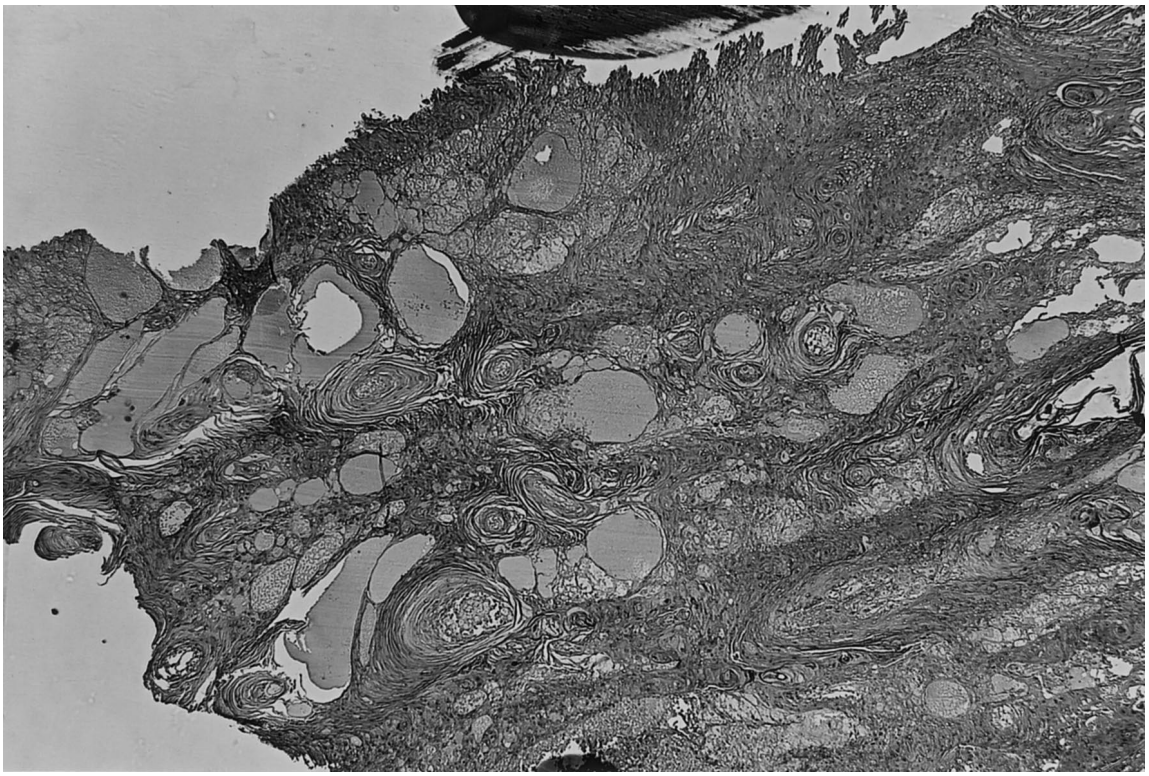

FIG. 1. Peritoneal implant displaying well-differentiated squamous cell carcinoma.

surgery uneventfully and was discharged from hospital the following day.

Pathologic analysis of the specimen from the right ovary revealed a right dermoid cyst and a benign left ovarian follicle. The right ovarian cyst measuring 5.7 by 4.8 by 1.8 cm weighed 30 g. The lumen of the cystic structure was filled with hair and sebaceous material. Microscopic examination indicated that several of the sections of the cyst contained foci of moderately differentiated infiltrating squamous cell carcinoma. The carcinoma appeared to be confined to the cyst. Other than the surgical site, there was no evidence of penetration through the cyst capsule.

Upon receipt of the pathology report, referral was made to a gynecologic oncologist. The patient was asymptomatic and the physical exam was unremarkable at the time of assessment, 20 days after the initial surgery. A computed axial tomography scan of the abdomen and pelvis was significant for a 4.0 by 2.5 cm mixed cystic and solid mass arising from the left ovary as well as a locally enlarged, irregular uterus. No pelvic lymphadenopathy was identified. A radiograph of the chest was normal.

Thirty-three days after the initial laproscopic surgery, a staging laparotomy was performed. The procedure consisted of a total abdominal hysterectomy and bilateral salpingo-oophorectomy, bilateral dissection of the obturator, common iliac, and para-aortic lymph nodes, and omentectomy. Biopsies were taken from areas of fibrinous exudate on the pelvic sidewalls. Obvious hair and sebaceous debris were visible in the pelvic cavity.

Pathologic analysis was remarkable for two specimens labeled “left anterior peritoneal nodule” and “cul-de-sac nodule” showing implants of a well differentiated squamous cell carcinoma (Fig. 1). The right ovary contained a residual parenchymal focus of atypical keratinocytes also consistent with squamous carcinoma. All lymph nodes and omentum were negative for cancer. There was a foreign body inflammatory reaction to implanted hair found in the resected omentum, the peritoneal biopsies, and both ovaries. The uterus contained multiple small fibroid tumors. Based on these findings, the patient was classified as having a FIGO stage IIc squamous cell carcinoma arising in an ovarian dermoid cyst.

Subsequent to surgery, the patient was treated with adjuvant chemotherapy concurrent with radiotherapy. She received five cycles of chemotherapy. The first, fourth, and fifth cycles consisted of cisplatin (40 mg/m^2) and etoposide (50 mg/m^2). During the second and third cycles she received cisplatin (60 mg/m^2) with concurrent pelvic and total abdominal radiotherapy for carcinoma of the ovary. First, a midplane dose of 2250 cGy of 10-MV photons was administered in 10 fractions of 225 cGy over 2 weeks via 17 by 17 cm anterior–posterior ports to the pelvis. Then, the abdomen and pelvis received a midplane dose of 2250 cGy of 6-MV photons in 22 fractions of 102 cGy over 4$\frac{1}{2}$ weeks via 44.6 by 28.0 cm anterior and 28.0 by 4.28 cm posterior ports. After the fifth cycle, the patient elected to

TABLE 1
Guidelines for Laparoscopic Management of Ovarian Masses [10, 11]

Risk factor for malignancy	Criteria
Personal or family history of ovarian cancer	None
Age of the patient	Premenopausal
Size of the mass	5 cm or less
Characteristics of the mass on ultrasound	Unilocular, unilateral, smooth border, lack of papillary excrescences, hypoechogenic or homogeneous (excluding endometriomas and dermoid cysts)
Tumor markers	Within normal range
Findings at diagnostic laparoscopy	Lack of dense adhesions, lack of papillary excrescences

discontinue chemotherapy due to the related side effects. She is currently free of disease after 36 months of follow-up.

DISCUSSION

The risk of dissemination of malignancy following rupture of a dermoid cyst is often minimized because malignancies occur only rarely in such cysts, especially in premenopausal women. Malignancy is found in association with dermoid cysts in only 1–2% of cases and there are no particular signs or symptoms which are characteristic of malignancy arising in a dermoid [3]. The frequency of malignant degeneration is related to age, with the highest incidence in the postmenopausal years [3]. Squamous cell carcinoma accounts for 70–88% of all malignant tumors arising in mature cystic teratomas [4]. The most common sites of spread at initial laparotomy are the pelvic peritoneum and the omentum [3, 4].

Squamous cell carcinoma arising in mature cystic teratomas has a poor prognosis when the disease has spread beyond the ovary [3]. Adequately staged patients with disease confined to the ovary have a much better prognosis with 5-year survival rates approaching 95% [5]. Although there is still no consensus on specific prognostic indicators for this disease, the grade and FIGO stage of the tumor, as well as the presence of residual disease or vascular invasion, have been shown to be predictive of survival in retrospective studies [5, 6].

The prognostic effect of rupture of the capsule with spillage and upstaging of the tumor from Ia to IIc remains controversial [7]. An important limiting factor in these studies is the often inadequate surgical staging in reported cases and the inconsistency of adjuvant treatments offered.

With regard to the current report, several things are cause for concern. The first is that the rupture that occurred was intentional. Deliberate rupture to facilitate cyst removal has been previously reported, although the removal of dermoid cysts with intact capsules or with minimal spill [2] (using an endobag, for example) has also been described. In a retrospective study, the rate of spillage was less than 4% when laparotomy was performed versus 100% when the dermoid was removed laparoscopically [1].

Whether intraoperative rupture of stage Ia ovarian cancer with subsequent upstaging of the tumor effects prognosis is still controversial. The time interval between laparoscopy and the eventual removal of the diseased ovary at laparotomy is a more important prognostic factor than intraoperative rupture of the cyst [7, 8].

Although some authors have suggested that early stage ovarian cancer may be treated laparoscopically, we feel that the finding of malignant disease at laparoscopy warrants laparotomy. Several cases of ovarian cancer spread and upstaging secondary to laparoscopic intervention have been reported [9]. This results in an important delay before staging laparotomy. The present case is the first to report upstaging of a carcinoma arising in a presumed benign teratoma secondary to deliberate intraoperative rupture during laparoscopic intervention.

Dermoid cysts of the ovary in young women pose a dilemma because of the ease of diagnosis on ultrasound and the small chance of associated malignancy. It has generally been considered acceptable to manage dermoid cysts laparoscopically in premenopausal women despite the associated high incidence of spillage [1, 2]. This case should serve to emphasize that malignant degeneration does occasionally occur in younger age groups, even when the cyst is fairly small, and that upstaging secondary to spillage at laparoscopy is a significant risk. Upstaging may worsen prognosis and almost certainly leads to more aggressive therapy that can impose significant morbidity on the patient.

Consent for laparoscopic removal of a dermoid cyst should consider the rare possibility of malignancy and the common possibility of spillage. A discussion of how spillage might affect prognosis may also be appropriate. Criteria (Table 1) are available for the selection of patients for laparoscopic management [10, 11]. Before dissecting the cyst, careful inspection of all peritoneal surfaces and pelvic organs should be done and peritoneal cytology performed (as would be done in a staging laparotomy). The cyst should be removed in an endobag to minimize the risk of spillage, and should spillage occur generous irrigation of the cavity should be performed. Obtaining quick sections if spillage occurs is warranted in order to eliminate the delay before staging laparotomy if the cyst's contents are malignant. Converting to laparotomy should always be considered a safe alternative rather than a defeat when the

surgeon has doubts about the feasibility of removing a cyst intact.

In conclusion, malignant degeneration within a dermoid cyst is rare in premenopausal women, but it certainly does occur. Spillage rates with laparoscopic removal are inevitably higher than with excision at laparotomy. In this case, spillage of dermoid contents at laparoscopy led to upstaging of a malignant tumor from stage Ia to IIc, possibly affecting prognosis. The upstaging led to aggressive adjuvant therapy which imposed significant morbidity, including loss of fertility, on the patient.

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